Spontaneous Closure of Cerebral Dural Arteriovenous Fistulas with Direct Cortical Venous Drainage

A Case Report

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Summary

We describe two rare cases of spontaneous closure of cerebral dural arteriovenous fistulas (DAVFs) with a small nidus and draining directly in a single cortical vein with several ectasias.

Eighteen previously published cases of spontaneous closure of cerebral DAVF comprised more benign fistula types. In literature, several explanations for DAVF occlusion have been proposed. We hypothesize that, in addition to the known causes, the specific contrast medium used during the diagnostic selective angiography might have played a role in the thrombosis and subsequent fistula closure.

Introduction

Cerebral dural arteriovenous fistulas (DAVFs) are rare cerebral vascular entities. Most DAVF show drainage to a dural sinus. Intracranial DAVFs with direct drainage to cortical veins only are classified as type III according to Borden et Al [1]. In the presence of accompanying venous ectasia, the DAVF is graded a type IV DAVF according to Cognard et Al [2]. Such intracranial DAVFs have a poor prognosis and need treatment because of the high risk of cerebral hemorrhage and neurological deficit [3,4].

Spontaneous closure of cerebral DAVF is incidentally reported and limited to DAVFs with drainage to a dural sinus with or without reflux into cortical veins, i.e. Borden and Cognard type I and II DAVF. The mechanism of spontaneous closure of DAVFs is unknown. We describe two cases of spontaneous closure of type III cerebral DAVFs, occurring shortly after diagnostic selective cerebral digital subtraction angiography (DSA).

Case Reports

Patient 1

A 53-year-old woman was admitted to an affiliated hospital in September 2005 with sudden headache, visual loss, aphasia and right-sided hemiparesis. Computed tomography (CT) scans revealed thrombosis of the left transverse sinus and dilated cortical veins, without bleeding or infarction. Anticoagulation therapy was instituted with subsequent full recovery of neurological symptoms. Follow-up magnetic resonance (MR) imaging in November 2005 showed complete occlusion of the left transverse and sigmoid sinus, and internal jugular vein and a fistula or AVM on the left side (Figure 1A). In February 2006, the patient was transferred to our hospital for DSA revealing a Cognard type IV DAVF with exclusive external carotid arterial supply by dural branches from the left middle meningeal and occipital arteries, converging in a small fistula and draining into a dilated osteodural vein showing venous ectasia. Additionally, retrograde leptomeningeal drainage into several dilated cortical cerebral veins was seen (Figure 1B). Several hours post angiography, the patient had sudden onset headache, aphasia and progressive right-sided hemiparesis. CT scan revealed no

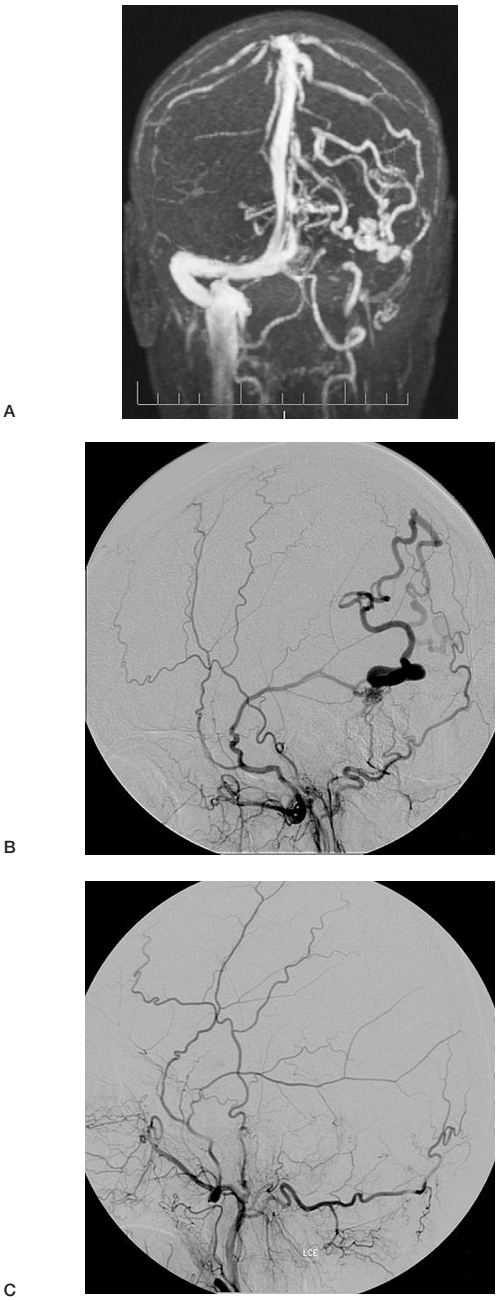

Figure 1 A 53-year-old woman presenting with headache, visual loss, aphasia and right sided hemi paresis. Maximum intensity projection MR angiography image (A) demonstrates a small fistula or AVM with dilated cortical veins of the left hemisphere and occlusion of the left sided transverse and sigmoid sinus. The clinical symptoms resolved gradually. Lateral left external carotid artery angiogram (B) reveals a DAVF supplied by dural branches from both the middle meningeal and occipital artery, a small nidus and direct drainage to a dilated osteodural vein showing venous ectasia and retrograde leptomeningeal drainage into several dilated cortical cerebral veins. Several hours after the cerebral DSA, the patient developed progressive clinical symptoms. Control angiogram of the left external carotid artery (C) confirmed the obliteration of the DAVF.

hemorrhage and subsequent MR imaging suggested occlusion of the DAVF. Control cerebral DSA in May 2006 (postponed by the patient out of fear) actually showed closure of the DAVF (Figure 1C). The patient recovered completely.

Patient 2

A 63-year-old woman underwent brain MR imaging in an affiliated hospital because of hearing problems and dizziness. As a coincidental finding an isolated dilated vessel structure was seen in the anterior left frontal lobe. The patient was referred to our hospital in March 2009 for cerebral DSA (Figure 2A,B) that disclosed a left sided Cognard type IV frontal fossa DAVF with multiple dural arterial feeders from the ethmoid branches of both ophthalmic arteries and secondary supply from the external carotid artery, converging in a single fistula at the cribriform plate bone and draining directly into a dilated osteodural vein with several venous ectasias, draining finally into the superior sagittal sinus and also retrograde in superficial cortical cerebral veins. Selective arterial endovascular treatment was proposed because of the high bleeding risk. Several days after angiography, the patient had sudden onset of left frontal headaches without neurological deficit. Pre-embolization cerebral DSA showed complete occlusion of the DAVF (Figure 2C). Thrombosis of the draining vein was confirmed by MR imaging (Figure 2D). The headache gradually subsided.

Discussion

Dural arteriovenous fistulas (DAVFs) are abnormal vascular shunts within the dura. Most frequently DAVFs develop near venous sinuses, but they can occur at any site within the dura. Arterial supply is usually from dural artery branches, mainly originating from the external carotid artery. Venous drainage occurs most frequently via dural sinuses, less often via osteodural veins or retrograde via leptomeningeal veins, or a combination. In adults, DAVF are acquired and are probably associated with venous (sinus) thrombosis. DAVF usually present with pulsatile tinnitus, less frequently with hemorrhage or neurological deficit. DAVFs with direct cortical venous drainage are uncommon and will quite often go undetected until they cause hemorrhages. Anterior fossa DAVFs are even more exceptional. Fistula closure happens only when the nidus is occluded. This can be obtained through selective endovascular occlusion or surgical resection. The treatment risk must be

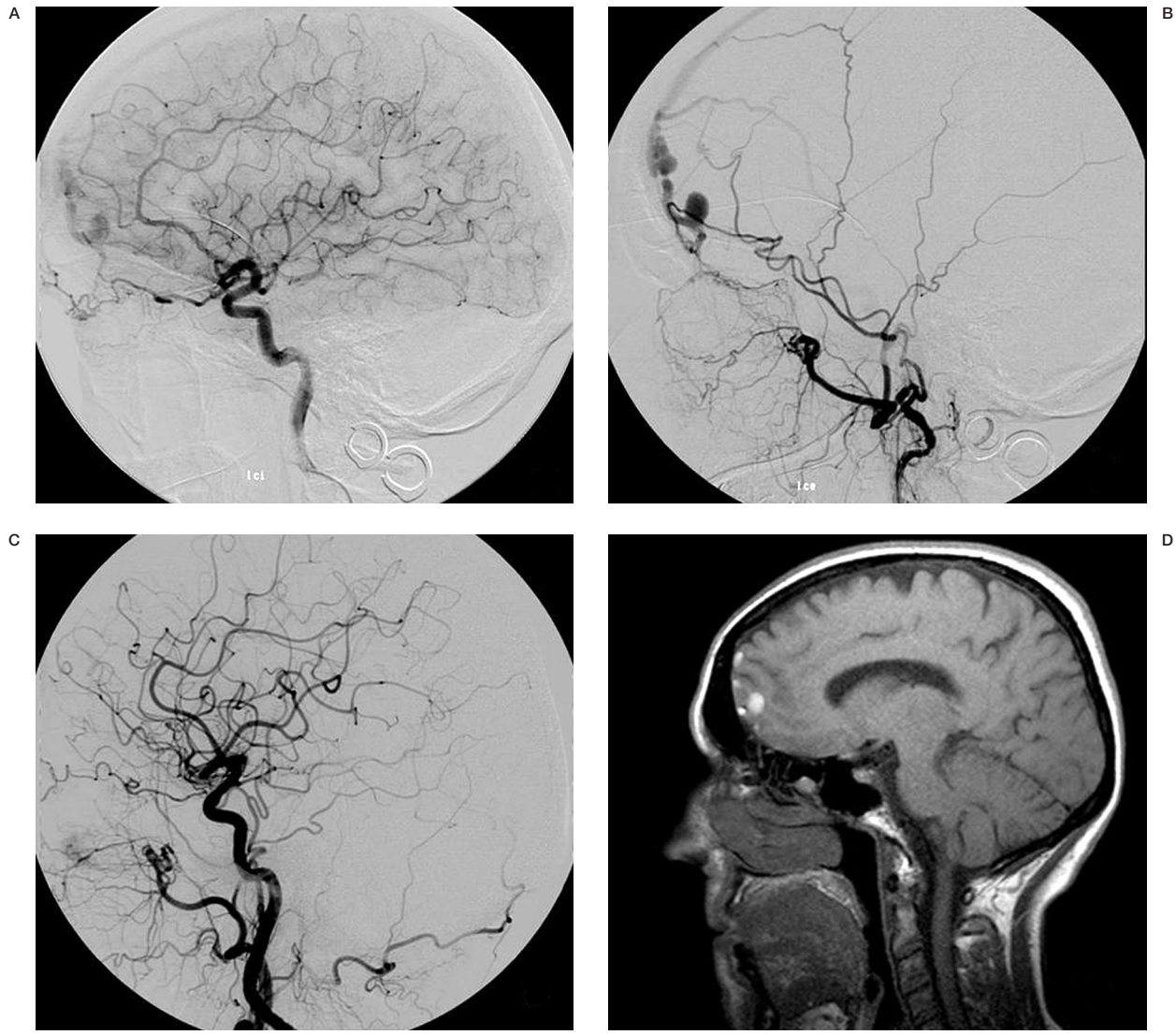

Figure 2 A 63-year old woman presenting with an incidental finding of an anterior fossa DAVF. Lateral left internal (A) and external (B) carotid artery angiogram reveals a DAVF supplied by dural ethmoidal branches from both ophthalmic arteries and also from dural branches from the external carotis arteries (mainly the anterior middle meningeal and anterior ethmoidal arteries), with a small nidus and direct filling of a dilated osteodural left frontal vein with several venous ectasias. Control angiogram (C) several weeks after the initial diagnostic DSA demonstrates the occlusion of the anterior fossa DAVF. MR imaging (D) confirms the thrombosis of the single draining cortical vein on sagittal T1 weighted images.

weighed against the natural course. Most Borden type III DAVFs have an aggressive clinical course and should be treated by a neurovascular team [3-5]. Literature review shows 18 previously described cases of spontaneous closure of cerebral DAVF. Like our cases, most DAVF developed spontaneously [6-10]. To our knowledge, spontaneous closure of a Borden type III or Cognard type IV intracranial DAVF have not been reported to date. The cause of spontaneous closure of DAVFs is unknown. Some authors have hypothesized that thrombosis of the involved dural sinus induced fistula occlusion; others proposed that associated intracranial hemorrhage produces mass-effect or secondary vasospasm of the feeding arteries [9]. Damage to vessels involved in posttraumatic DAVF could lead to scar tissue and subsequent spontaneous closure [6]. DAVFs with small fistulas with single draining veins may lead to total venous outflow obstruction and lesion thrombosis, as in our cases [6,9]. Intracranial hemorrhage at initial presentation, a small nidus and the presence of a sin-

gle draining vein seem to play a major role in spontaneous regression of pial brain arteriovenous malformations [11-14]. In our cases a small fistula and single draining vein were present, but no posttraumatic intracranial hemorrhage was seen.

Fistula closure may be triggered by the cerebral DSA to map the DAVF [9,10]. Selective feeding artery catheterization may result in decelerating shunt flow because of temporary blood flow stagnation. However, in our cases no vasospasm was present and the fistulas remained patent on all control angiograms. More likely, gradual progressive thrombo-occlusion may be caused by thrombogenic effects of the contrast medium used, especially in slow-flow shunts consisting of a small nidus draining into a single vein. The contrast medium used in our angiographies was Omnipaque® 300 (GE Healthcare, CO/ Cork, Ireland), a low osmolar, non-ionic contrast medium containing 647 mg of the active substance Iohexol per milliliter. Iohexol induces arterial endothelial changes in morphology with no effect on fibrin deposition [15]. Formed thrombi will be larger and more resistant to thrombolysis compared to saline controls [16]. In patients with DAVFs or other arteriovenous malformations in the presence of a small fistula or nidus and a single draining vein, thrombo-occlusion of the fistula or nidus might occur shortly after the DSA, possibly partly induced by the contrast medium used.

Conclusions

We describe two rare cases of spontaneous closure of cerebral DAVFs with a small nidus draining directly into a single dilated cortical vein with several ectasias in its course. Eighteen previously published cases of spontaneous closure of cerebral DAVF comprised more benign fistula types, involving a dural sinus. In literature, several explanations for DAVF occlusion have been proposed. We hypothesize that, in addition to the known causes, the specific contrast medium used during the cerebral DSA might have played a role in the thrombosis and subsequent fistula closure.

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